



City Research Online

City, University of London Institutional Repository

Citation: van den Hoek -Engel, L., Harding, C., van Gerven, M. & Cockerill, H. (2017). Pediatric feeding and swallowing rehabilitation: An overview. *Journal of Pediatric Rehabilitation Medicine*, 10(2), pp. 95-105. doi: 10.3233/PRM-170435

This is the accepted version of the paper.

This version of the publication may differ from the final published version.

Permanent repository link: <https://openaccess.city.ac.uk/id/eprint/17752/>

Link to published version: <https://doi.org/10.3233/PRM-170435>

Copyright: City Research Online aims to make research outputs of City, University of London available to a wider audience. Copyright and Moral Rights remain with the author(s) and/or copyright holders. URLs from City Research Online may be freely distributed and linked to.

Reuse: Copies of full items can be used for personal research or study, educational, or not-for-profit purposes without prior permission or charge. Provided that the authors, title and full bibliographic details are credited, a hyperlink and/or URL is given for the original metadata page and the content is not changed in any way.

Journal of Pediatric Rehabilitation Medicine: An
Interdisciplinary Approach 10 (2017) 95–105 95
DOI 10.3233/PRM-170435

IOS Press

Pediatric feeding and swallowing rehabilitation: An overview

Lenie van den Engel-Hoek^a;₁, Celia Harding^b, Marjo
van Gerven^a and Helen Cockerill^c

*a: Donders Centre for Neuroscience, Department of
Rehabilitation, Radboud University Medical Center, Nijmegen,
The Netherlands*

b : City University, the Royal Free Hospital, London, UK

*c: Evelina London Children's Hospital, Guy's and St. Thomas's
NHS Foundation Trust, London, UK*

Accepted 20 January 2017

Abstract. Children with neurological disabilities frequently have problems with feeding and swallowing. Such problems have

a significant impact on the health and well-being of these children and their families. The primary aims in the rehabilitation of pediatric feeding and swallowing disorders are focused on supporting growth, nutrition and hydration, the development of feeding activities, and ensuring safe swallowing with the aim of preventing choking and aspiration pneumonia. Pediatric feeding and swallowing disorders can be divided into four groups: transient, developmental, chronic or progressive. This article provides an overview of the available literature about the rehabilitation of feeding and swallowing disorders in infants and children. Principles of motor control, motor learning and neuroplasticity are discussed for the four groups of children with feeding and swallowing disorders.

Keywords: *Pediatric, rehabilitation, feeding and swallowing, dysphagia*

1. Introduction

The act of feeding and swallowing is typically described in three phases. In the oral phase (suckling during breast feeding, sucking when bottle feeding, eating with a spoon, drinking from a cup or chewing solid food) the bolus is prepared and transported to the pharynx. In the pharyngeal phase the swallow reflex is triggered and the bolus is moved through the pharynx. In the esophageal phase the bolus is

transported to the stomach. Swallowing is a complex process that involves reflexive and voluntary motor control and intraoral sensory processing. Together with sensorimotor integration and autonomic regulation it enables the safe ingestion of food and liquid [1]. Since 2007 the American Speech-Language-Hearing Association documents have adopted 'feeding and swallowing disorders' as the more inclusive phrase for dysphagia and delays and/or disorders in the development of eating and drinking skills, which are common in a variety of pediatric populations [2,3]. Feeding and swallowing disorders have a great impact on the health and well-being of children and their families. Dysphagia can also have a negative effect on dietary intake, growth and development [4]. In addition, parents experience stressful child-caregiver interaction, worries about choking and poor growth, and social isolation [5,6].

Feeding and swallowing disorders vary widely in terms of presentation, severity and frequency, and the available treatment strategies [7]. As discussed by Arvedson et al. (2010), the disorders can be divided into four different groups: transient, developmental, chronic or progressive [7]. In examples where the skills have not been acquired within the typical age range, due to prematurity, neurological conditions or developmental delay, specialized training strategies and more practice may be needed for the child to acquire feeding skills [8]. In children with progressive diseases or trauma, adaptation and compensatory strategies need consideration.

The primary aims in the rehabilitation of pediatric feeding and swallowing disorders are focused on supporting nutrition and hydration, adequate growth, on the development of feeding skills, and ensuring safe swallowing, as well as the prevention of choking and aspiration pneumonia. Pediatric swallowing rehabilitation must be considered as different from adult swallowing rehabilitation. In children the oropharyngeal anatomy is still developing, oral movements mature from reflexive to volitional and children learn to manage a range of food textures as well as learning to use a variety of different utensils. Infants develop from being totally dependent during feeding to totally self-feeding. In contrast, the swallowing rehabilitation in adults depends on a memory for motor components of swallowing (often referred to as an 'internal model') [9], whereas in infants and children new skills have to be learned. Therefore, the term habilitation is sometimes used. Habilitation and rehabilitation both focus on the act of learning skills. The difference between the two is that habilitation focuses on learning new skills whereas rehabilitation focuses on regaining skills that have been lost. In this overview, we will use the term rehabilitation to represent both habilitation and rehabilitation due to the overlap between transient, developmental, chronic and progressive disorders of feeding and swallowing.

Clinicians who treat children with feeding and swallowing problems frequently incorporate oral-motor exercises (OME) into their treatment plans. In the light of motor learning concepts and principles of task specificity (see next paragraph for description of these terms), controversy exists about the theoretical basis and effectiveness of these interventions for individuals with swallowing disorders, particularly in pediatric populations [7]. Consequently, there has been a shift towards therapy goals which are based on motor learning for feeding and swallowing skills during the mealtime itself [7,8,10].

The aim of this paper is to provide an overview of the available recent literature about the rehabilitation of feeding and swallowing disorders in infants and children, and the available evidence on a range of interventions. The article will include: (1) a description of the principles of motor control and motor learning and the role in the development of feeding and swallowing; (2) an overview of the available evidence for the use of oral motor exercises in pediatric dysphagia rehabilitation; and (3) the current evidence related to the treatment of the four main patient groups of feeding and swallowing disorders. Examples of rehabilitation based on the concepts of motor learning will be given.

2. Motor control and motor learning

Motor control and motor learning are essential components of the development of feeding and swallowing in children [8]. Motor control is the process of creating a sequence of movements for the performance of coordinated and skilled actions. This involves interaction between the central and peripheral nervous systems. The importance of sensory input for accurate motor control and motor learning must be emphasized. Food of different consistencies initiate swallowing by taste, touch, temperature and pressure. Increased sensory input can modify motor areas of the cerebral cortex [11]. Stimulation of a greater number of receptive fields induces a stronger reflex with greater muscle activity and force [12]. Ease of movement is possible due to a steady stream of sensory information while an activity is planned, executed and evaluated [9]. In addition to using sensory feedback during an activity, the human nervous system also has the capacity to predict sensory consequences of motor commands. Shadmehr and colleagues (2010) concluded that human beings are able to estimate the movement required by predicting what should happen, and by receiving information of the sensory system about what did happen, to make comparisons [13]. These concepts are significant for oropharyngeal swallowing. It is suggested that while a bolus is still in the oral cavity, some sensory predictions about readiness for the pharyngeal swallow are made, related to bolus size, temperature, and consistency.

In infants and children all these activities have to be learned through experience. Motor learning and motor adaptation are important in the development of new feeding skills. Motor learning is defined as the acquisition of skills or skilled movements as a result of practice. It is a gradual process based on reduction of error and the development of successful movements. Sensory information is used in a feedback and feedforward way to modify motor activity [9]. For example in the acquisition of chewing, different stages can be identified. Animal studies have shown that tactile receptive fields on the tongue often have reciprocal receptive fields on food, seen in the initial stages of eating chewable food, is not only a primitive way of eating, but has the important function of learning the sensory characteristics of solid food, such as consistency and flavor. Gagging, which is often seen when solid food is introduced, can be seen as a warning: this piece is too big to swallow [15]. Using this information infants will act differently with the next bite. Motor learning, with the influence of sensory information with feedback and feedforward, will help them to swallow an increasing range of food textures effectively. Neuroimaging studies have demonstrated a strong link between motor learning and neuroplasticity [16]. Neuroplasticity refers to the ability of the brain to change and to adapt to new conditions. Change and adaptation occur, for example, in response to training, experience and aging. These changes lead to behavioural changes [17]. Principles of neuroplasticity (such as 'age matters', 'specificity', 'use it or lose it', 'repetition and age matters', and 'transference') and motor learning related to swallowing rehabilitation in adults are important to consider [17]. Attempts have been made to translate some of the principles to pediatric swallowing rehabilitation (Table 1) [18]. The principle of 'age matters' refers to the well-recognized idea that training influences more readily the younger system than the older nervous system. Task-specific training is the repetitive practice of a task that is specific to the intended outcome [8]. The principle of 'specificity' suggests that a treatment exercise should closely parallel the desired task [19,20]. 'Use it or lose it' is a concept which is important in the treatment of progressive diseases. Although principles such as 'repetition matters' and 'intensity matters' are thought to be significant in rehabilitation, it is not known what and how much is effective in pediatric feeding and swallowing rehabilitation. The 'transference' principle highlights the idea that tasks that are learned easily can be performed in other circumstances. In general it is accepted that an early start of intervention ('age matters') and task specificity are important [18,21]. Although not specific to children with dysphagia, principles of motor learning can provide guidance to clinicians in selecting appropriate therapeutic strategies when only limited or equivocal research is available.

3. Active and passive oral motor exercises

Oral-motor exercises (OME) are often incorporated into treatment plans for children with feeding and swallowing disorders. There are three main categories of OME generally used in clinical practice: active exercises, passive exercises, and sensory approaches. Active exercises include an active range of movement (such as sticking out of the tongue) and strength training (such as exercises for tongue strengthening). These exercises are aimed at increasing strength, endurance, and power. Sjogreen and colleagues (2010) used lip strengthening exercises in children with myotonic dystrophy and found increased lip strength, but no generalisation to improved functional feeding and swallowing [22]. This well designed study underlines the necessity to use task specific training in children with feeding and swallowing disorders. The evidence base for the use of OME in isolation, outside a functional setting, is weak. The assumed relation between OME and the activity of eating and swallowing is questionable [10]. It is likely that different sites in the central nervous system are being activated during nutritive movements and during volitional motor activities that are often trained with OME [23]. Arvedson and colleagues (2010) found in their systematic review, that there is insufficient evidence to determine the effects of OME on the skills of children with oral sensorimotor deficits and swallowing problems [7]. Passive exercises may include massage, stroking, stretching, and tapping, with little active movement from the individual receiving treatment [7,24,25]. These procedures are applied to provide sensory input or improve circulation. It has been theorized that some of these techniques normalize feeding patterns by reducing abnormal oral reflexes, facilitating normal muscle tone and normal movements or desensitizing the oral region. However, benefits of the passive exercises have not been reported [26]. The typical stretch reflexes, which can be seen in skeletal muscles are lacking in tongue and lip muscles [26,27]. Therefore it is unlikely that these exercises would improve oral strength or alter muscle tone.

Sensory approaches usually consist of the application of heat, cold, electrical stimulation, high frequency vibration or other activity involving stimulation of the skin and muscle tissues. Although common in the past, many of these techniques are no longer used. They were based on the idea that vibration or icing and brushing would influence muscle activity over a long period (20–30 minutes) [28]. However, studies have shown that the effect only lasted for 30–45 seconds [29]. These studies contributed to a change towards task specific training and therapy based on motor learning in therapy for feeding and swallowing disorders by speech language therapists (SLTs) [29].

Neuromuscular electrical stimulation (NMES) of the anterior neck muscles has been explored in the treatment of adults who have dysphagia. The hypothesis is that the electrical impulses can improve pharyngeal muscle strength in combination with accelerated cortical reorganization [30]. Chen and colleagues (2016) found in their review that the evidence was insufficient to indicate that neuromuscular electrical stimulation alone was superior to swallow therapy in dysphagia after stroke [31]. NMES has been tested in a group of children who had dysphagia with a variety of disorders, both congenital and acquired [30]. The authors tested NMES treatment of anterior neck muscles, compared to a similar group who received traditional therapy approaches to remediate dysphagia. Patients who received NMES did not show improved swallow function in comparison with patients who received traditional therapeutic interventions. Both groups were heterogeneous and there is a possibility that there could be subgroups of children that will improve with NMES treatment, especially those with acquired dysphagia. Further studies could perhaps consider this.

In addition, swallowing manoeuvres, such as the supraglottic swallow, effortful swallow or the Mendelsohn manoeuvre have largely been tested in the adult population and not with children. Morgan

stated that their application should be limited to older children who have adult-like swallowing physiology [18].

In summary, the evidence for the benefits of OME in pediatric dysphagia rehabilitation is weak. Recent studies show the theoretical advantages of the use of principles of motor learning in pediatric feeding and swallowing rehabilitation. Humbert (2013) showed the combined influence of motor learning and oral sensory feedback on the cortical representations of oral muscles [9]. SLTs are increasingly incorporating these principles in their treatment, underlining the need to perform well designed studies in this field.

4. Pediatric feeding and swallowing disorders in the four patient groups

4.1. Transient feeding and swallowing disorders

Transient feeding and swallowing disorders are frequently seen in children who were born prematurely. A significant amount of research has been performed in the last two decades to describe the feeding and swallowing disorders, to investigate possible treatment and to assess the outcome of treatment. The prevalence of feeding problems in former premature infants is twice that of full term infants and the reasons for feeding problems are multifactorial [32]. SLTs are often involved in the assessment and management of feeding in this population. Keeping in mind the concepts of motor learning and task specificity, knowledge of normal development of sucking, swallowing and breathing is essential in the management of this patient group. In addition, the pharyngo-esophageal motility and airway protection mechanisms and the overall state of infants have to be taken into account. Jadcherla and colleagues (2012) have described an interesting approach incorporating all these elements [32]. The cue-based feeding approach in oral feeding therapy, described in this study, refers to individualized, developmentally appropriate practice instead of the traditionally volume-driven feeding model. In the initial phase non-nutritive sucking (on a pacifier) to stimulate sucking and prepare for nutritive sucking is provided [33]. However, it should be noted that non-nutritive sucking does not always enable an infant to achieve full oral feeding more quickly than those infants who do not use non-nutritive sucking [34]. Using non-nutritive sucking is important for helping the infant to achieve the quiet alert state appropriate for feeding, and it can support parent learning on how to interpret differing infant states and therefore support parent-bonding [34]. After the initial phase, oral feeding is introduced in a stepwise manner, at around the age of 34 weeks gestational age when coordination of sucking, swallowing and breathing is neurodevelopmentally possible [35]. The importance of observing the infant before, during and after feeding, is emphasized by the Early Feeding Scale [36]. This instrument supports the observation of the impact of sucking and swallowing on respiratory rate, heart rhythm and oxygen saturation. It enables the neonatal practitioner to observe how the infant's overall neurological system is maturing in relation to feeding. From research it is known that premature born infants need well-coordinated sucking, swallowing and breathing to cope with milk intake. However, this pattern may not have been established when oral feeding is started [37]. Initial feeding attempts at 32 to 34 weeks are characterized by uncoordinated suck-swallow-breathe sequences, with some apnoeic episodes. As the infant develops, the suck-swallow-breathe cycle becomes more coordinated with longer sequential suck bursts, shorter pauses between sucks, and fewer apnoeic episodes. Finally, breathing efforts are integrated into an overall suck-swallow-breath rhythm around 36 weeks [35]. The initial series of apnoeic suckle runs can have a negative influence on feeding experience because of choking and desaturation. In line with the principles of task specific training, pacing can be used in combination with a low flow teat. Pacing involves the feeder regulating the number of sucks per burst and the duration of bursts and pauses by systematically removing the nipple from the mouth, or interrupting the seal on the nipple [38]. By using pacing the neonate is offered early experiences with oral feeding ('age matters') without negative experiences.

Parents are encouraged to feed their child with pacing to train safe oral feeding and enhance the relation with their child. In addition, pharmacological management in case of bronchopulmonary dysplasia or gastroesophageal reflux is essential and must be based on assessment and evidence based guidelines. The overall goal of treatment is to improve the quality of the feeding experience for both the infant and parent rather than, in the first place, focusing on the quantity of food intake [3].

4.2. Developmental feeding and swallowing disorders

Developmental feeding and swallowing disorders are seen in children with developmental delay or with genetic syndromes as a result of the complex interactions between anatomical, medical, physiological and behavioural factors [39]. For example, in children with Down's syndrome a combination of problems might be the cause of dysphagia. Developmental delay and neuromotor incoordination, in combination with hypotonia, poor tongue control and open mouth posture, often interfere with the acquisition of effective feeding skills. The management of these feeding difficulties is often possible with an appropriate feeding program (matching with the mental and/or developmental stage of the child) which consists of three important factors.

A feeding program that (1) must be safe, (2) must support optimal growth and nutrition, and (3) must be realistic and based on the development of feeding skills. Normal developmental acquisition of eating skills in infants is described in terms of milestones [8]. The broad milestones are considered to be nipple (breast or bottle), eating from a spoon, drinking from a cup, and biting and chewing [40]. Weaning is the process of expanding the diet of an infant to include food and drinks other than breast milk or formula [41]. Feeding in the weaning period is considered not only as the transition from milk to solids, but also the transition from sucking to eating with a spoon, and to chewing and biting. Furthermore, the interaction between caregiver and child is gradually replaced by independent eating and drinking by the child. The child's new skills are gradually improved by experience in combination with the use of different utensils and a variety of foods. For example, the first attempts by the child to remove the food from the spoon may be uncoordinated, but through trial and error and with repetitions during daily feeding sessions the new skill is acquired in 5–7 weeks [15]. In this motor learning process, efficiency and independence through daily, functional experiences and practice are progressively achieved as the skill improves. In children with developmental delay and difficulty with feeding, it will take more time to improve this skill in combination with careful rehabilitation. Thelen (1989) suggests that it is the task, and not any pre-existing practice of component skills, that supports the development of the emerging skill [42]. She considers both action and sensory perception, as represented in the target task, important in learning new motor skills. These should not be replaced by simulated tasks in which component skills are practiced, functional skill. Therefore, eating from a spoon has to be learned, whilst food of an appropriate texture is presented on a spoon. Therapy interventions should provide practice with gradually increased bolus size, number of accepted spoons, and promptness in taking the food [8].

Problems with chewing are often reported in children with developmental disabilities and are very complex, involving the growing oral anatomy, the sensory information which has to be combined with adequate motor responses, and the experience of new consistencies and tastes. The oral anatomy consisting of bones, muscles, teeth and soft tissues is dynamic over the course of a child's development. Changes occur during growth, but are also affected by the activity of chewing. For example, palatal width and height increase extensively in the first two years, requiring modification of the motor planning during chewing [43]. The development of chewing efficiency starts with munching and crushing and ends in efficient chewing with lateral movements of the tongue followed by the movement of the food from the teeth to the pharyngeal area for swallowing [43]. The effect of food consistency on children's development of chewing has not been explored widely. Both human and animal studies have reported

the effect of food consistency on orofacial development, suggesting that a diet of soft food might negatively influence bone and muscle growth and therefore reduce chewing competence [44]. This underlines the necessity of paying attention to the process of mastication in children with developmental delay in the early stages of life. To learn the skill of effective chewing, specificity and repetition are the most important components. Insufficient mastication is often accompanied by frequent gagging, because small children try to swallow pieces that are too large, and inadequately prepared.

A protocol with carefully described steps can positively impact on the development of chewing solid foods. This should include chewing skills that are trained by first placing foods on the preferred side for chewing, enhancing graded jaw movement and lateral tongue movements. Food texture is carefully modified, working from soft to mixed textures and then later to firm consistencies. Practicing mastication in different situations highlights the 'transference principle', because of the influence of the first experiences on the performance of the skill in a variety of environments [8]. During this process, motor control, sensory tolerance and sensory capabilities are developing simultaneously resulting in the new skill of mastication [8].

4.3. Chronic feeding and swallowing disorders

Infants and children with neurological disorders often have dysphagia, based on the insufficient neural coordination of swallowing, alongside other problems such as hypertonicity or hypotonicity. Cerebral palsy (CP) is the most common form of neurodevelopmental disability. Prevalences of dysphagia range from 43% to 99% in children with CP of varying levels of physical disability [45,46]. In CP dysphagia is often characterized by problems in both the volitional oral movements and the more reflexive pharyngeal phase of swallowing.

Moreover, impaired ability to plan and coordinate swallowing with respiration is often present, due to the central nervous system disorder. This lack of coordination can result in aspiration (with cough, or silent) with aspiration pneumonia, often being reported [47]. With changes in nutritional needs (i.e. related to the requirement of increased quantities of food and a broader range of consistencies, which might be too difficult to swallow), growth of the oropharyngeal area in puberty, increased scoliosis and increasing spasticity, aspiration may become more frequent in older children [47]. Arvedson and colleagues (1994) identified that in children with CP, aspiration was most prominent for liquid, either alone or with some other texture [48]. In these patients, aspiration does not appear to be an isolated event. It is highly associated with disorders of pharyngeal and esophageal motility. First attempts have been made to describe these problems in children by Rommel and her research group [49]. In children with CP they used the deglutitive flow interval, derived from pharyngeal impedance recordings. The impedance measurements can detect alterations in flow characteristics of pharyngeal swallowing resulting in post swallow residue. This flow interval was clearly altered in relation to pathology and may potentially be combined with pressure measurement as a measure of dysfunction. It highlights the complexity of dysphagia at all stages of swallowing in children with CP. It would seem logical that feeding and swallowing interventions lead to benefits, which are measurable and objective. However, the current level of evidence is poor with limited information regarding outcomes. Randomized controlled trials for intervention are difficult in this heterogeneous population of patients with CP, with ethical concerns about not intervening in children who have an impaired swallow [47]. Only 5 randomized controlled trials were found in a review of 12 electronic databases [50]. Feeding safety and efficiency were primary outcomes in some studies, height and weight changes in others. Various interventions are described in children with CP: (1) oral sensorimotor

programs and/or muscle strengthening, (2) thickening liquids, (3) changing postures and (4) the use of motor learning in CP. Gisel (1994) completed a study in children with CP which explored the development of oral motor skills [51]. She compared training of oral motor skills (referred to as oral sensorimotor therapy) in different groups of children with CP. Specific skills such as lip closure for the retention of food, and biting to break through a piece of solid food were trained with small food bits, before mealtime. Lip closure, biting and chewing improved after 20 weeks. Although lip closure improved, drinking from a straw or cup did not improve, which could be explained in terms of task specific learning. Oral motor exercises and muscle strengthening training have not been shown to be effective in promoting feeding efficiency or weight gain in children and adolescents with CP suggesting that strengthening interventions in isolation are neither effective nor worthwhile [50]. Traditionally, in case of aspiration on thin liquids, thickened liquids are recommended. If we consider the data from the Arvedson and colleagues study (1994), dysphagia in CP is not only characterized by aspiration on thin liquid, but residue after swallow is often observed with the possibility of indirect/delayed aspiration [48]. In a study on the videofluoroscopic swallowing recordings of 112 children with neurologic conditions (CP and neuromuscular disorders) direct aspiration on thin liquid in the CP group was observed. However, indirect/delayed aspiration was additionally seen as a result of pharyngeal post swallow residue with pureed food [52]. Thickening liquids poses practical problems for caregivers and research has shown that thickened liquids are difficult to mix precisely. Although thickness or viscosity is often the focus of measurement, other material property characteristics of the internal structure of the liquid, such as density and yield stress, affect the way that thickened liquids move and behave [53]. The International Dysphagia Diet Standardization Initiative (www.IDDSI) has recently developed global standardized terminology and definitions for texture modified foods and thickened liquids to be used with individuals with dysphagia. The terminology could also be helpful for clinical practice in infants and children.

Head and trunk stability, in combination with alignment of oral structures, are important in feeding and swallowing [54]. The postural difficulties of children with CP challenge therapists and parents to establish the optimal positioning for feeding. Hyperextension of the neck may hamper laryngeal protection during swallowing [55]. It is hypothesized that positioning of the trunk and head is the most basic and essential treatment for dysphasic children with CP. Snider et al. (2010) found in their systematic review that there is only limited evidence (based on a few case series) that positioning has a positive effect on feeding and swallowing in CP children [50]. In a small study with 6 children it was shown that with a slightly reclined posture and a flexed neck, aspiration decreased both on thin liquids and pureed food [55]. The effects of motor learning and task specificity in CP were investigated by Pinnington and Hegarty (2000), through a consistent method of food presentation [56]. The commonly used posture for children with CP, positioned in the midline with neck flexion during feeding, was combined with an often used recommendation given to caregivers that food should be presented on a spoon with a relatively flat bowl, without scraping the utensil against the teeth. In the study this was all achieved with a robotic arm that the children could control themselves. Statistically significant differences in components of oral motor behavior between the assessment periods were found with this consistent method of food presentation, which could not be attributed to maturation alone. This small study showed the benefits of the training a specific oral motor behavior during feeding. It supports the idea that also in CP children, with the use of clearly described advices and techniques, motor learning for specific tasks could be beneficial.

4.4. Progressive feeding and swallowing disorders

In children and young adults with neuromuscular disorders (NMD) or muscle diseases, such as spinal muscular atrophy (SMA), myotonic dystrophy or Duchenne muscular dystrophy (DMD) feeding

problems and dysphagia may develop slowly. In children with NMD malnutrition, impaired chewing performance and oral phase problems, choking and post swallow residue are described [57]. The common element in all NMDs is muscle weakness, which influences motor abilities and oral motor activities for feeding and swallowing, being influenced by the specific muscle groups affected by the NMD. The oral phase of swallowing can be affected, resulting in drooling of saliva, losing food out of the mouth, mastication problems, piecemeal deglutition, poor bolus formation, and need more time to complete their meals due to these oral phase problems. Pharyngeal phase problems can range from residue after swallow in the valleculae, piriform sinuses and pharyngeal wall to penetration of food above the vocal folds or aspiration, more with solid food than with thin liquid [52]. NMDs are often complicated by oral motor and structural anomalies, which may negatively influence the oral phase of swallowing. A wide range of structural difficulties are reported, such as malocclusions, limited mouth opening, tented upper lip, high arched palate, an atrophic or hypertrophic tongue, and dystrophic oral muscles [58–61]. In DMD oral muscles have been visualized and dystrophic changes were quantified using muscle ultrasound [62]. It has been shown that the range of movement of the oral muscles gradually decreased over time, reflecting the increasing influence of structural dystrophic changes. The echogenicity of the oral muscles showed the same gradual involvement as observed in various skeletal muscles, but at a later onset. These dystrophic changes in skeletal and oral muscles were found to be related to muscle weakness [62,63]. The oral muscle weakness in this patient group causes more problems with thick liquid and solid food. Rehabilitation in these patient groups must focus on compensatory strategies and low intensity training. Compensatory strategies are described in terms of modifying consistencies, i.e. smaller pieces of chewable food, less solid food and more thin liquid, and modifying posture. Children with SMA frequently have a retracted head posture. Adjusting the head to a more upright position can lead to less pharyngeal post swallow residue and a better swallowing when eating solid food [64].

The early involvement of masticatory muscles in DMD is an explanation for chewing problems and an open mouth posture in early stages of the disease [65]. From other studies with DMD it is known that disuse leads to deterioration, and research has shown that a low-intensity physical training is beneficial in terms of preservation of muscle endurance and functional abilities [66]. This principle was extrapolated to chewing and a study with mastication training was performed with the use of one piece of sugar-free chewing gum per exercise (3 _ 30 min/day, 5 days/week, 4 weeks long). The masticatory performance was assessed using a mixing ability test and the anterior bite force was measured. The masticatory performance in the intervention group improved and the improvement remained after the one-month follow-up [67]. Bite force did not improve. The specific task of mastication was trained and improved, again highlighting the benefits of task specific training. It is important to realize that different progressive neuromuscular disorders will follow a specific trajectory in terms of the involvement of oral muscles, resulting in different signs and symptoms of dysphagia. Therefore knowledge of dysphagia and its different presentations with pediatric NMDs is important to enable SLTs to design tailored interventions to suit individual needs [68].

5. Conclusions

Knowledge about pediatric feeding and swallowing and the management of dysphagia have greatly improved during the past two decades. Normal motor development of feeding and swallowing may be disrupted by a sudden deficiency in a particular skill resulting in inefficient or unsafe feeding and swallowing. The development may also be delayed, e.g. in children with neurodevelopmental disabilities, or children may gradually experience more feeding and swallowing problems in case of a progressive disease [39]. Growth, nutrition, hydration, prevention of aspiration pneumonias and quality of life should always be the main goals in feeding and swallowing therapy. In recent years the treatment of feeding

and swallowing disorders in children with developmental disabilities has increasingly focused on using motor learning approaches. Studies have resulted in a shift from using oral motor exercises, aimed at improving component skills, to task specific training during functional everyday mealtimes with the focus on motor learning to minimize risks associated with feeding and swallowing disorders. Functional training should be used to improve motor skills [42]. The best practice for eating is the act of eating itself. This contrasts with a bottom-up approach in which individual motor skills, such as jaw, lip and tongue exercises, are trained in the expectation they will be combined and generalized to the task of eating. So, for practicing feeding skills one might argue: “a child learns to eat with a spoon when it is actually fed with a spoon”. An understanding of the concepts of motor control and motor learning, as well as knowledge of the normal development of feeding and swallowing, the presenting problems in different patient groups, the influence of food properties (i.e. differences between thin and thick liquid), and of oralthe pharyngeal phase has become important for SLTs working with children and young people with feeding and swallowing disorders.

Conflict of interest

All authors declare no conflict of interest.

References

- [1] Rangarathnam B, Kamarunas E, McCullough GH. Role of Cerebellum in Deglutition and Deglutition Disorders. *Cerebellum*. 2014; 13: 767-76. doi: 10.1007/s12311-014-0584-1.
- [2] Homer EM. Management of Swallowing and Feeding disorders in Schools. https://www.pluralpublishing.com/media/media_msfds_SamplePages.pdf; 2006 [cited 2016].
- [3] American Speech-Language-Hearing Association A. Pediatric Dysphagia. 2016 [cited Retrieved 02-20-2016, from <http://www.asha.org/Practice-Portal/Clinical-Topics/Pediatric-Dysphagia>].
- [4] Dodrill P, Gosa MM. Pediatric Dysphagia: Physiology, Assessment, and Management. *Ann Nutr Metab*. 2015; 66 Suppl 5: 24-31. doi: 10.1159/000381372.
- [5] Lefton-Greif MA, Okelo SO, Wright JM, Collaco JM, McGrath-Morrow SA, Eakin MN. Impact of children’s feeding/ swallowing problems: validation of a new caregiver instrument. *Dysphagia*. 2014; 29(6): 671-7. doi: 10.1007/s00455-014-9560-7.
- [6] Craig GM. Psychosocial aspects of feeding children with neurodisability. *Eur J Clin Nutr*. 2013; 67 Suppl 2: S17-20. doi: 10.1038/ejcn.2013.226.
- [7] Arvedson J, Clark H, Lazarus C, Schooling T, Frymark T. Evidence-based systematic review: effects of oral motor interventions on feeding and swallowing in preterm infants. *Am J Speech-Lang Pat*. 2010; 19(4): 321-40. doi: 10.1044/1058-

0360(2010/09-0067).

[8] Sheppard JJ. Using motor learning approaches for treating swallowing and feeding disorders: a review. *Lang Speech Hear Ser.* 2008; 39(2): 227-36.

[9] Humbert IA, German RZ. New directions for understanding neural control in swallowing: the potential and promise of motor learning. *Dysphagia.* 2013; 28(1): 1-10. doi: 10.1007/s00455-012-9432-y.

[10] Harding C, Cockerill H. Managing eating and drinking difficulties (dysphagia) with children who have learning disabilities: What is effective? *Clinical Child Psychology and Psychiatry.* 2015; 20(3): 10. doi: 10.1177/1359104513516650.

[11] Gow D, Hobson AR, Furlong P, Hamdy S. Characterising the central mechanisms of sensory modulation in human swallowing motor cortex. *Clin Neurophysiol.* 2004; 115(10): 2382-90. doi: 10.1016/j.clinph.2004.05.017.

[12] Steele CM, Miller AJ. Sensory input pathways and mechanisms in swallowing: a review. *Dysphagia.* 2010; 25(4): 323-33.

[13] Shadmehr R, Smith MA, Krakauer JW. Error correction, sensory prediction, and adaptation in motor control. *Annu Rev Neurosci.* 2010; 33: 89-108. doi: 10.1146/annurev-neuro-060909-153135.

[14] Sweazey RD, Bradley RM. Response characteristics of lamb pontine neurons to stimulation of the oral cavity and epiglottis with different sensory modalities. *J Neurophysiol.* 1993; 70(3): 1168-80.

[15] van den Engel-Hoek L, van Hulst K, van Gerven M, van Haaften L, de Groot S. Development of oral motor behavior related to the skill assisted spoon feeding. *Infant Behav Dev.* 2014; 37(2): 187-91. doi: 10.1016/j.infbeh.2014.01.008.

[16] Dayan E, Cohen LG. Neuroplasticity subserving motor skill learning. *Neuron.* 2011; 72(3): 443-54. doi: 10.1016/j.neuron.2011.10.008.

[17] Robbins J, Butler SG, Daniels SK, Diez Gross R, Langmore S, Lazarus CL, et al. Swallowing and dysphagia rehabilitation: translating principles of neural plasticity into clinically oriented evidence. *J Speech Lang Hear Res.* 2008; 51(1): S276-300. doi: 10.1044/1092-4388(2008/021).

[18] Morgan AT. Management of oromotor disorder for feeding in children with neurological impairment. In: Roig-Quilis MP, L., editor. *Oromotor disorders in childhood.* Barcelona: Viguera Editores, L.S.; 2011.

[19] Maas E, Robin DA, Austermann Hula SN, Freedman SE, Wulf G, Ballard KJ, et al. Principles of motor learning in treatment of motor speech disorders. *Am J Speech-Lang Pat.* 2008; 17(3): 277-98. doi: 10.1044/1058-0360(2008/025).

[20] Wilson EM, Green JR, Yunusova Y, Moore CA. Task specificity

in early oral motor development. *Semin Speech Lang.* 2008; 29(4): 257-66. doi: 10.1055/s-0028-1103389.

[21] Kleim JA, Jones TA. Principles of experience-dependent neural plasticity: implications for rehabilitation after brain damage. *J Speech Lang Hear Res.* 2008; 51(1): S225-39. doi: 10.1044/1092-4388(2008/018).

[22] Sjogreen L, Tulinius M, Kiliaridis S, Lohmander A. The effect of lip strengthening exercises in children and adolescents with myotonic dystrophy type 1. *Int J Pediatr Otorhi.* 2010; 74(10): 1126-34. doi: 10.1016/j.ijporl.2010.06.013.

[23] Kent RD. The uniqueness of speech among motor systems. *Clin Linguist Phonet.* 2004; 18(6-8): 495-505.

[24] Ottenbacher K, Hicks J, Roark A, Swinea J. Oral sensorimotor therapy in the developmentally disabled: a multiple baseline study. *Am J Occup Ther.* 1983; 37(8): 541-7.

[25] Beckman D. Beckman oral motor interventions, course pack accompanying Oral Motor Assessment and intervention Workshop. Charlotte, NC: 2001 August 2001.

[26] Clark HM. Neuromuscular treatments for speech and swallowing: a tutorial. *Am J Speech Lang Pathol.* 2003; 12(4): 400-15. doi: 10.1044/1058-0360(2003/086).

[27] Ruscello DM. Nonspeech oral motor treatment issues related to children with developmental speech sound disorders. *Lang Speech Hear Serv Sch.* 2008; 39(3): 380-91. doi: 10.1044/0161-1461(2008/036).

[28] Rood M. The use of sensory receptors to activate, facilitate and inhibit motor response, autonomic and somatic, in developmental sequence. In: Sattely C, editor. *Study course VI. Approaches to the treatment of patients with neuromuscular dysfunction.* Dubuque, Iowa: Brown & Co; 1962.

[29] Galea MP. Physical modalities in the treatment of neurological dysfunction. *Clin Neurol Neurosurg.* 2012; 114(5): 483-8. doi: 10.1016/j.clineuro.2012.01.009.

[30] Christiaanse ME, Mabe B, Russell G, Simeone TL, Fortunato J, Rubin B. Neuromuscular electrical stimulation is no more effective than usual care for the treatment of primary dysphagia in children. *Pediatr Pulmonol.* 2011; 46(6): 559-65. doi: 10.1002/ppul.21400.

[31] Chen YW, Chang KH, Chen HC, Liang WM, Wang YH, Lintion on post-stroke dysphagia: a systemic review and metaanalysis. *Clin Rehabil.* 2016; 30(1): 24-35. doi: 10.1177/0269215515571681.

[32] Jadcherla SR, Peng J, Moore R, Saavedra J, Shepherd E, Fernandez S, et al. Impact of Personalized Feeding Program in 100 NICU Infants: Pathophysiology-based Approach for better outcomes. *Hepatology and Nutrition.* 2012; 54(1): 62-71.

[33] Mizuno K, Ueda A. Changes in sucking performance from nonnutritive sucking to nutritive sucking during breast- and

- bottle-feeding. *Pediatr Res*. 2006; 59(5): 728-31.
- [34] Harding C, Frank L, Van Someren V, Hilari K, Botting N. How does non-nutritive sucking support infant feeding? *Infant Behav Dev*. 2014; 37(4): 457-64. doi: 10.1016/j.infbeh.2014.05.002.
- [35] Gewolb IH, Vice FL. Maturation changes in the rhythms, patterning, and coordination of respiration and swallow during feeding in preterm and term infants. *Dev Med Child Neurol*. 2006; 48(7): 589-94.
- [36] Thoyre SM, Shaker CS, Pridham KF. The early feeding skills assessment for preterm infants. *Neonatal Netw*. 2005; 24(3): 7-16.
- [37] Lau C, Smith EO, Schanler RJ. Coordination of suck-swallow and swallow respiration in preterm infants. *Acta Paediatr*. 2003; 92(6): 721-7.
- [38] Law-Morstatt L, Judd DM, Snyder P, Baier RJ, Dhanireddy R. Pacing as a treatment technique for transitional sucking patterns. *J Perinatol*. 2003; 23(6): 483-8. doi: 10.1038/sj.jp.7210976.
- [39] Cooper-Brown L, Copeland S, Dailey S, Downey D, Petersen MC, Stimson C, et al. Feeding and swallowing dysfunction in genetic syndromes. *Dev Disabil Res Rev*. 2008; 14(2): 147-57.
- [40] Arvedson JC. Assessment of pediatric dysphagia and feeding disorders: clinical and instrumental approaches. *Dev Disabil Res Rev*. 2008; 14(2): 118-27.
- [41] Northstone K, Emmett P, Nethersole F, Pregnancy ASTALSo, Childhood. The effect of age of introduction to lumpy solids on foods eaten and reported feeding difficulties at 6 and 15 months. *J hum Nutr Diet*. 2001; 14(1): 43-54.
- [42] Thelen E. The (re)discovery of motor development: Learning new things from an old field. *Dev Psychol*. 1989; 25(6): 4.
- [43] Le Reverend BJ, Edelson LR, Loret C. Anatomical, functional, physiological and behavioural aspects of the development of mastication in early childhood. *Brit J Nutr*. 2014; 111(3): 403-14. doi: 10.1017/S0007114513002699.
- [44] Ciochon RL, Nisbett RA, Corruccini RS. Dietary consistency and craniofacial development related to masticatory function in minipigs. *J Craniofac Genet Dev Biol*. 1997; 17(2): 96-102.
- [45] Erasmus CE, van Hulst K, Rotteveel JJ, Willemsen MA, Jongerius PH. Clinical practice : Swallowing problems in cerebral palsy. *Eur J Pediatr*. 2012; 171: 409-14.
- [46] Calis EA, Veugelers R, Sheppard JJ, Tibboel D, Evenhuis HM, Penning C. Dysphagia in children with severe generalized cerebral palsy and intellectual disability. *Dev Med Child Neurol*. 2008; 50(8): 625-30. doi: 10.1111/j.1469-8749.2008.03047.x.
- [47] Arvedson JC. Feeding children with cerebral palsy and swallowing

difficulties. *Eur J Clin Nutr.* 2013; 67 Suppl 2: S9-12.
doi: 10.1038/ejcn.2013.224.

[48] Arvedson J, Rogers B, Buck G, Smart P, Msall M. Silent aspiration prominent in children with dysphagia. *Int J Pediatr Otorhi.* 1994; 28(2-3): 173-81.

[49] Noll L, Rommel N, Davidson GP, Omari TI. Pharyngeal flow interval: a novel impedance-based parameter correlating with aspiration. *Neurogastroent Motil.* 2011; 23(6): 551-e206. doi: 10.1111/j.1365-2982.2010.01634.x.

[50] Snider L, Majnemer A, Darsaklis V. Feeding interventions for children with cerebral palsy: a review of the evidence. *Physical & occupational therapy in pediatrics.* 2011; 31(1): 58-77. doi: 10.3109/01942638.2010.523397.

[51] Gisel EG. Oral-motor skills following sensorimotor intervention in the moderately eating-impaired child with cerebral palsy. *Dysphagia.* 1994; 9(3): 180-92.

[52] van den Engel-Hoek L, Erasmus CE, van Hulst KC, Arvedson JC, de Groot IJ, de Swart BJ. Children with central and peripheral neurologic disorders have distinguishable patterns of dysphagia on videofluoroscopic swallow study. *J Child Neurol.* 2014; 29(5): 646-53. doi: 10.1177/0883073813501871.

[53] Cichero JA, Steele C, Duivesteyn J, Clave P, Chen J, Kayashita J, et al. The Need for International Terminology and Definitions for Texture-Modified Foods and Thickened Liquids Used in Dysphagia Management: Foundations of a Global Initiative. *Current Physical Medicine Rehabilitation Reports.* 2013; 1: 280-91. doi: 10.1007/s40141-013-0024-z.

[54] Redstone F, West JF. The importance of postural control for feeding. *J Pediatr Nurs.* 2004; 30(2): 97-100.

[55] Larnert G, Ekberg O. Positioning improves the oral and pharyngeal swallowing function in children with cerebral palsy. *Acta Paediatr.* 1995; 84(6): 689-92.

[56] Pinnington L, Hegarty J. Effects of consistent food presentation on oral-motor skill acquisition in children with severe neurological impairment. *Dysphagia.* 2000; 15(4): 213-23.

[57] Tilton AH, Miller MD, Khoshoo V. Nutrition and swallowing in pediatric neuromuscular patients. *Semin Pediatr Neurol.* 1998; 5(2): 106-15.

[58] Kiliaridis S, Katsaros C. The effects of myotonic dystrophy and Duchenne muscular dystrophy on the orofacial muscles and dentofacial morphology. *Acta Odontol Scand.* 1998; 56(6): 369-74. doi.

[59] Granger MW, Buschang PH, Throckmorton GS, Iannaccone ST. Masticatory muscle function in patients with spinal muscular atrophy. *Am J Orthod Dentofacial Orthop.* 1999; 115(6): 697-702.

[60] Sjogreen L, Engvall M, Ekstrom AB, Lohmander A, Kiliaridis S, Tulinius M. Orofacial dysfunction in children and

- adolescents with myotonic dystrophy. *Dev Med Child Neurol*. 2007; 49(1): 18-22.
- [61] Wang CH, Finkel RS, Bertini ES, Schroth M, Simonds A, Wong B, et al. Consensus statement for standard of care in spinal muscular atrophy. *J Child Neurol*. 2007; 22(8): 1027-49.
- [62] van den Engel-Hoek L, Erasmus CE, Hendriks JC, Geurts AC, Klein WM, Pillen S, et al. Oral muscles are progressively affected in Duchenne muscular dystrophy: implications for dysphagia treatment. *J Neurol*. 2013; 260(5): 1295-303. doi: 10.1007/s00415-012-6793-y.
- [63] Jansen M, van Alfen N, Nijhuis-van der Sanden MWG, van Dijk JP, Pillen S, de Groot IJM. Quantitative muscle ultrasound is a promising longitudinal follow-up tool in Duchenne Muscular Dystrophy. *Neuromuscul Disord*. 2012; 22(4): 306-17.
- [64] van den Engel-Hoek L, Erasmus CE, van Bruggen HW, de Swart BJ, Sie LT, Steenks MH, et al. Dysphagia in spinal muscular atrophy type II: more than a bulbar problem? *Neurology*. 2009; 73(21): 1787-91.
- [65] van den Engel-Hoek L, de Groot IJ, Sie LT, van Bruggen HW, de Groot SA, Erasmus CE, et al. Dystrophic changes in masticatory muscles related chewing problems and malocclusions in Duchenne muscular dystrophy. *Neuromuscul Disord*. 2016; 26(6): 354-60. doi: 10.1016/j.nmd.2016.03.008.
- [66] Jansen M, van Alfen N, Geurts ACH, de Groot IJM. Assisted bicycle training delays functional deterioration in boys with Duchenne muscular dystrophy: the randomized controlled trial "no use is disuse". *Neurorehabil Neural Repair*. 2013; 27(9): 816-27. doi: 10.1177/1545968313496326.
- [67] van Bruggen HW, van den Engel-Hoek L, Steenks MH, Bilt AV, Bronkhorst EM, Creugers NH, et al. Fighting Against Disuse of the Masticatory System in Duchenne Muscular Dystrophy: A Pilot Study Using Chewing Gum. *J Child Neurol*. 2015; 30(12): 7. doi: 10.1177/0883073815575575.
- [68] van den Engel-Hoek L, de Groot IJ, de Swart BJ, Erasmus CE. Feeding and Swallowing Disorders in Pediatric Neuromuscular Diseases: An Overview. *J Neuromuscul Dis*. 2015; 2(4): 357-69. doi: 10.3233/JND-150122.